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TRAP (Twin Reversed Arterial Perfusion) sequence

Case Report

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Abstract: TRAP (Twin Reversed Arterial Perfusion) sequence is a rare malformation that occurs in monozygotic twin pregnancies. Twin weight ratio in our case was 76% (body weight of pump twin was 1800 g, acardiac twin 1370 g), but in spite of that the pump twin had a mild clinical picture with premature birth, fetal hydrops (mild pleural effusion; hypoproteinemia; on the 3rd day of life the baby was without edema and the level of his serum proteins were in referral ranges) and mild myocardial hypertrophy of left ventricle (echocardiography performed on the 28th day of life was normal).

Keywords: *Acardiac twin • Pump twin • Hydrops fetalis*

1. Introduction

TRAP (Twin Reversed Arterial Perfusion) sequence is a rare malformation that occurs in monozygotic twin pregnancies. As a consequence of abnormal blood supply from structurally normal, pump twin through placental anastomoses, normal development of the heart and other organs is missing in the other one (acardiac/recipient twin). Pump twin provides circulation for himself as well as for the acardiac one over reversed circulation through great arterio-arterial and veno-venous anastomoses. Fetal physiology is thereby severely disturbed with consecutive increase of perinatal mortality. Most of pump twins are without congenital malformations, although in about 10% of cases some of them can be found, such as congenital heart defects, gastroshisis and skeletal anomalies. One of the possible explanations is that in pump twin hemodynamic imbalance occurs with fluctuations of blood pressure and possible blood loss of the pump twin to a cardiac one [1].

Various types of acardiac twin could develop: the *aa rdiac ae phalus* – absent cephalic development; the *aa rdiac ane ps* – cranial structures and/or neural tissue are present; the *aa rdiac ao rmus*–fetus has cephalic structures, but poorly developed or totally absent thorax; the *aa rdiac amorphous* – neither cephalic nor thoracic structures have been developed. In our case the acardiac acephalus type has been found [2].

The incidence of TRAP sequence is approximately 1% of monochorionic twin pregnancies or 1 in 35.000-150.000 deliveries [2].

2. Case report

A 26 year-old woman, G2P1 with no significant medical history attended our department at 21 weeks gestation. She was sent by her local doctor with a diagnosis of placental tumour and excessive abdominal enlargement. Ultrasound examination revealed monochorionic

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monoamniotic twin pregnancy. The first twin was normal and corresponding to gestational age and a second acardiac twin that was acephalus and acardiac with associated hydramnios. An amorphous mass with gross edema was measuring 11x8 cm with reversed arterial blood flow (Figure 1). The parents were counseled about the possibility of TRAP sequence and the prognosis for the surviving co-twin. Cordocentesis was performed and revealed a normal male karyotype. The patient was seen at every two weeks for ultrasound examination, fetal echocardiography and Doppler ultrasound, umbilical artery (UA) pulsatility index (PI) and middle cerebral artery (MCA) PI and peak systolic velocity (Vmax). A follow up scan showed that the surviving twin was growing well and the acardiac twin has increased in size. Patient was admitted at 29 weeks of gestation and due to fetal anaemia of the pump twin delivered at 30+5/7 weeks gestation by elective caesarean section. Amniotic fluid was clear, polyhidramnios. Acardiac twin weight was 1370 g. The Apgar scores of our patient were 4, 1 and 7 at 1, 5 and 10 minutes, respectively. His body weight was 1800 g (75 percentile), body length 42 cm (50 percentile), head circumference 31 cm (91 percentile). After initial resuscitation, due to bradicardia (heart rate 90/min.) baby was intubated and assisted ventilation has been started. He was transferred on mechanical ventilation to referral neonatal intensive care unit of our Institute.

On admission, male preterm newborn in the 3rd hour of life, heart rate 140/min, respiratory rate 40/min, oxygen saturation 94%. Severe generalized subcutaneous edema, bilaterally diminished respiratory sounds, mild heart murmur (II/VI). Axial and segmental hypotonia.

Complete blood count, blood gasses and other biochemical parameters, except serum proteins – 24g/l (normal value 45-55 g/l) were in referral ranges. TORCH and Parvovirus B19 (IgM) were negative. Karyotype: 46, XY, normal. Chest radiograph showed signs of respiratory distress syndrome of the second degree. Pleural ultrasound revealed minimal effusion in left frenicocostal sinus. Brain and abdominal ultrasound were normal. The electrocardiography was normal. Echocardiography on admission showed myocardial hypertrophy of left ventricle without blood flow obstruction. Echocardiography performed on the 28th day of life was normal.

Figure 1. Amorphous mass on antenatal ultrasound at 28 weeks of gestations

After admission, mechanical ventilation and standard parenteral antimicrobial therapy (ampicillin, gentamycin) were initiated. Because of hypoproteinemia and edema, fresh frozen plasma and Henley loop diuretics were given. On the 3rd day of life, the baby was with stable vital parameters, without edema and the level of his serum proteins were in referral ranges. He was extubated and transferred to neonatology unit. He was discharged on the 41st day of his life, in good condition and recommended for rehabilitation treatment because of central hypotonia.

3. Discussion

TRAP sequence is also called chorioangiopagus parasiticus. In utero, it was described for the first time in 1978 by Lehr and Dire [3]. Nowadays, it is usually diagnosed antenatally. By using modern 3D ultrasonography and color Doppler technology, condition can be determined as early as in the first trimester [4].

Pathogenesis is still unknown. One of the possible explanations is that arterial blood is deoxigenated as a result of reversed perfusion with consecutive disturbance of normal organ morphogenesis. According to another hypothesis, primary embriopathy is the cause of abnormal heart development followed by provision of blood from the pump twin to acardiac one through the great arterio-arterial and veno-venous anastomoses. This leads to reversal blood flow in umbilical blood vessels of acardiac twin. Contrary to regular fetal circulation, oxygenated blood runs to the fetus by an umbilical artery and deoxygenated blood from him by the umbilical vein. As a result of disturbed intrafetal circulation, the caudal part of malformed twin gets blood reacher in nutrients and oxygen than upper one and this situation leads to the better development of the pelvis and lower extremities. Completely desaturated blood runs retrogradely in the upper portion of the body and head with consequent disturbance of the heart and thoracic development that are wholly absent or severely deformed [5,6].

After TRAP sequence is diagnozed antenatally, it is of the utmost importance to secure the best possible outcome of structurally normal twin. Congestive heart failure, polyhydramnios and premature birth are possible complications in pump twin. Moore et al. described 49 cases of acardiac twins and determined that better

perinatal outcome could be expected in smaller twin weight ratios. If twin weight ratio is above 70%, previously mentioned complications are more frequent. In some instances, as in our patient, fetal hydrops can develop in pump twin. Perinatal mortality in pump twins is about 55% [7,8]. The weight (in grams) of the acardiac twin is calculated with the following formula: (1.2 x longest lenght2) – (1.7 x longest lenght) [9].

Twin weight ratio in our case was 76%, but inspite of that the pump twin had mild clinical picture with premature birth, fetal hydrops (mild pleural effusion; hypoproteinemia; on the 3rd day of life the baby was without edema and the level of his serum proteins were in referral ranges) and mild myocardial hypertrophy of left ventricle (echocardiography performed on the 28th day of life was normal).

In order to reduce or prevent possible complications in morphologically normal twin, repeated amniocentesis for reduction of amniotic fluid volume could be executed. Also, maternal digitalization for treatment of pump twins` congestive heart failure or selective premature birth of acardiac twin can be done. Lastly, one of therapeutic options is to stop perfusion of acardiac twin with selective bipolar umbilical cord cauterization, umbilical cord laser coagulation or endoscopic ligation [10].

Intrafetal ablation has also been performed by alcohol injection, monopolar diathermy, interstitial laser, and radiofrequency ablation (RFA). Many authors consider that the intrafetal RFA ablation is pretty safe, and minimally invasive and the most successful method. Livington and al. did the RFA threatment on 17 pregnant women who were diagnosed with the TRAP sequence. The average gestation age (GA) at time of the threatment was 21 weeks (17-24 weeks), and the average GA at the delivery was 37 weeks (26-39 weeks). In 95% of cases, the pump twin has survived [11]. The same rate of survival of the pump twin was achieved by Tsao and al [12].

On the other hand, Tan and Seuplveda recommend the intra-fetal ablation as a threatment of choice [13]. Hecher and al. found that the fetoscopic laser coagulation of placental anastomosis of the umbilical cord of the acardiac twin was successful in 82% of cases if only laser was used, and additional 15% if the laser was combined with the bipolar forceps. The success rate was higher if the threatment was applied in early second trimester [14].

TRAP sequence is a rare complication of monochorionic multiple gestation, but still it is very important to do an early and exact antenatal diagnostics as it might improve the outcome of this rare clinical entity. The accent is put on the minimum invasive threatments for the vascular anastomosis [15].

Conflict of interest statement

Authors state no conflict of interest.

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